



# Giant solitary hydatid cyst of spleen—A case report



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## ABSTRACT

**INTRODUCTION:** Primary hydatid disease of the spleen is very rare and even rarer to cause (any complication . . . pancreatitis.). Usually, splenic hydatid cysts are secondary, either resulting from spontaneous spread of cysts or occurring after operations involving hydatidosis in other regions. Here, we report a case of a primary isolated splenic hydatid cyst treated with a classical surgical approach. This case report and literature review describes the management of hydatid splenic invasion.

**PRESENTATION OF CASE:** We report the case of a 28-year-old female who presented with left hypochondriac non tender swelling/(mass). Abdominal ultrasonography and computed tomography (CT) revealed a cyst located in the spleen. The diagnosis was confirmed by a serological test. Surgical treatment involved a radical en bloc splenic resection (together with resection of the diaphragm and subcutaneous tissue.) The postoperative course was uneventful with three weeks of albendazole treatment. CT follow-up at six months demonstrated the absence of recurrence. Histopathologic examination revealed a hydatid cyst.

**DISCUSSION:** Complete aggressive surgical en bloc resection is the gold standard treatment of patients with hydatid cysts with the aim to remove all parasitic and pericystic tissues.

**CONCLUSION:** The infrequency with which it is encountered makes splenic hydatid disease a formidable early diagnostic challenge especially in nonendemic areas. Hydatid disease should be considered in the differential diagnosis of all cystic masses in the spleen/(abdomen), especially in the geographical regions where the disease is endemic.

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## 1. Introduction

Hydatid disease (Echinococcosis) is a zoonotic infection caused by the larval form of parasites of tapeworm, *Echinococcus granulosus*. Humans are the accidental intermediate host in the development cycle of hydatid disease. It is an endemic disease in the sheep and cattle raising countries Middle East, North Africa, New Zealand, Australia, and South America. Although hydatid disease affects any organ or soft tissue, it most frequently found in liver (60–70%), lungs (30%), and rarely encountered in the kidney, spleen, bone, thyroid, breast and pancreas. Clinical presentation varies according to the anatomic location of the cyst. Most hydatid cysts are acquired in childhood but a latent period of five to twenty years occurs before the diagnosis is made. The growth of hydatid cyst remains indolent yet unremitting by character. As a very crude estimate hydatid cysts increase their diameter by about two to three centimetres each year. The rate of growth of hydatid appears to be dependent on immunologic relationship between the parasites and humans as also on the resistance offered by the enveloping structure. Splenic hydatid disease is very rare with its occurrence less than 3% of the total incidence of Echinococcosis even in endemic areas. Berlot first described Splenic hydatid cyst as an autopsy

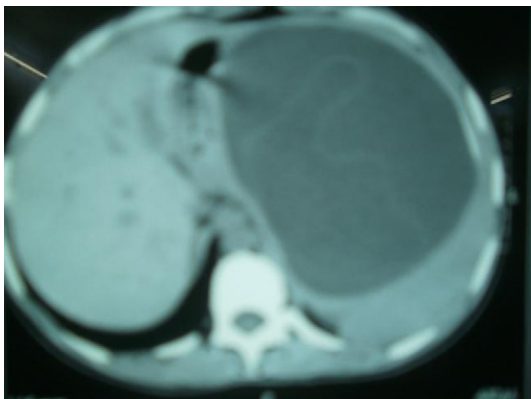
finding in 1790(1). Primary infestation of the spleen usually takes place by the arterial route after the parasite has passed the two filters (hepatic and pulmonary). A retrograde venous route, which bypasses the lung and liver, is also reported. Some complications may be seen with splenic hydatid cyst such as secondary infection, fistulisation to adjacent organs and rupture into the peritoneal cavity. The traumatic or spontaneous rupture of a hydatid cyst may cause a life-threatening complication of Systemic anaphylactic reaction.

### 1.1. Case presentation

A 28 years female patient farmer by occupation presented to our general surgery clinic with a mass in the left upper quadrant of the abdomen. There was left hypochondriac dull aching pain which did not shift or radiate. The size of lump increased rapidly over past 6 months. Patient complained of malaise with nausea, vomiting and weight loss since one year. Also there was intermittent fever every fifteen days since last six months. She had no history of jaundice, cough or respiratory distress, abdominal trauma, weight loss and her past medical history was unremarkable. On examination, her vital parameters were within normal limits. Physical examination showed an asymmetric abdomen and a growing lump with smooth surface in left hypochondriac, epigastric and umbilical region. Mild epigastric tenderness was noted with no rebound tenderness, guarding, or hepatomegaly. There was

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**Fig. 1.** Figure one CT scan picture shows large hydatid cyst in spleen, with normal liver, kidney and pancreas.

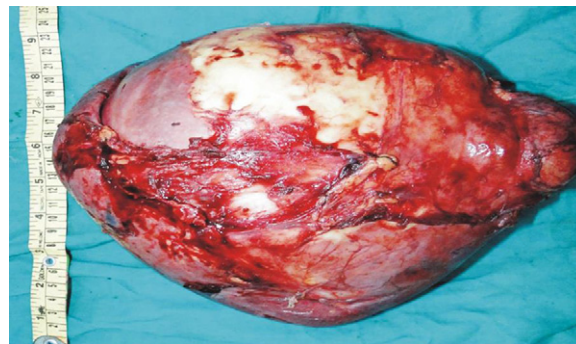
no lymphadenopathy. Chest, cardiovascular, central nervous, and the musculoskeletal systems were normal on examination.

Routine laboratory investigation CBC, coagulation profile, biochemistry, renal function test, liver function test and electrolytes revealed no abnormalities. ESR was of 75 mm/h (Westergren). Plain radiograph of the abdomen revealed a well-defined, rounded soft-tissue opacity with calcified margins in the left hypochondrium. Chest radiograph was normal. Abdominal ultrasonography showed round, well defined, cystic lesion of approx., size 165 × 140 mm over pancreas which moving left kidney and spleen.

Abdominal CT scan shows large homogenous cystic lesion in spleen measuring 20 × 22 cm loculated cyst with many septa, originating from the spleen. The cyst in the spleen appeared to fill the left quadrant of the abdominal cavity, displacing the intestines to the right, most likely suggestive of hydatid cyst. There were no cysts in other abdominal viscera. A CT scan of his chest did not show any cysts. Laparotomy was performed through a midline incision. Surgical exploration revealed a hydatid cyst occupying whole splenic parenchyma only thin rim of splenic tissue was present in inferior surface. The mass measuring approximately 250 × 200 mm was attached to left diaphragm. The cyst was resected en-bloc with the spleen. The rest of the abdominal organs including the liver were normal. In order to treat any potential contamination, the abdomen was washed locally with hypertonic saline solution (NaCl 20%). Histopathological examination showed the classic laminated cyst wall encircling many scolices with a double layer of hooklets; which is consistent with *Echinococcus granulosus* infection thus confirmed the diagnosis of splenic hydatid cyst. On cut section there was hydatid sand and fluid around 3.9 litres. The postoperative period was uneventful and the patient was discharged on the postoperative day 7. The postoperative course was uneventful with three additional weeks of albendazole treatment. The clinical and ultrasonography follow-up did not show any evidence of recurrence at six months (Figs. 1–3).

## 2. Discussion

Hydatid disease is a major health problem worldwide, mainly in sheep- and cattle-raising areas of the world. Hydatid disease of spleen is a rare clinical condition as even in the endemic region the frequency is reported to be 0.5–4% of abdominal hydatid diseases. The most common sites of hydatid disease are the liver (60–70%), which acts as a first filter and the lungs (10–40%), which acts as second filter. The rare sites include spleen, thyroid, gall bladder, central nervous system, kidney, psoas sheath, retroperitoneal region, orbit. Practically any organ can be infested by hydatid disease. Splenic involvement is uncommon event because cyst embryos are trapped in the liver and lungs, with only 15% entering systemic



**Fig. 2.** Gross picture of splenic hydatid cyst 20 cm × 25 cm.

circulation. The eggs of parasite escape the liver-lung barrier and cause Primary infestation of spleen through the arterial route 4. Splenic hydatid disease may also arise with retrograde spread of parasites via the portal and splenic veins bypassing the lung and liver. Secondary splenic hydatid disease usually follows systemic disseminated or intraperitoneal spread following ruptured hepatic hydatid cyst. The hydatid cyst consists of three layers. The outermost adventitia (pseudo cyst) is formed of compressed splenic tissue, a middle layer laminated membrane of friable ectocyst and an innermost germinal layer, endocyst. The presentation of splenic hydatid disease can vary greatly. Splenic hydatid cysts are usually asymptomatic, solitary slowgrowing and incidentally diagnosed. The main symptoms associated with the disease are abdominal discomfort, pain and palpable mass in the left upper quadrant. Tarcoveanu E. reported 38 cases of splenic Echinococcosis and abdominal pain was the most common symptom among these patients.<sup>2</sup> The complications of untreated splenic hydatid cyst are mainly secondary infection, inflammation (Lippit et al., 1967), and acute abdomen, compression of other viscera, intraabdominal rupture and fistulization to the bowel, mainly colon causing severe pain and may prove to be life-threatening pericystic inflammation may cause adhesions with nearby organs such as the kidney, left diaphragm, colon, stomach. Teke et al.<sup>3,4</sup> reported a splenic hydatid cyst perforating into the left colon and causing massive gastrointestinal bleeding. Rupture of splenic hydatid cyst into the thorax leading to spleno-thoracic fistula has also been reported. Severe anaphylactic reactions due to rupture of the cyst are also reported leading to fever, pruritus, dyspnoea, stridor and oedema of the face. Haematochezia may results from evacuation of the splenic pulp from the infarcted spleen into the colon. Growth may cause compression of the segmentary vessels of the spleen, which results in extensive pericystic splenic atrophy and the hydatid cyst may entirely replace the splenic parenchyma. The main differential diagnosis of splenic hydatidosis are cystic lesions of spleen such



**Fig. 3.** Shows cut section of splenic hydatid cyst in centre there is hydatid sand and fluid around 3.9 litres.

as splenic abscesses, epidermoid cysts, hematomas, post-traumatic pseudo cyst neoplasms like lymphangioma and haemangioma. Pre-operative diagnosis may be difficult due to the similarity of the presenting symptoms and the radiological findings to those of other more commonly encountered lesions of the spleen. Eosinophilia may be the finding on haematological investigation. The Casoni skin test is sensitive but not specific. Radiological diagnosis by plain X-Ray, Ultrasonography (USG), CT and MRI can also be used to diagnose hydatidosis. On abdominal or chest radiograph, marginal or crumpled eggshell-like calcifications in the splenic area are suggestive of splenic hydatidosis. Ultrasonography and computed tomography are the major diagnostic tools for splenic hydatid cyst. Serological tests are highly sensitive and specific for Echinococcosis. Given the limited efficacy of drug therapy and owing to the risk of spontaneous or traumatic rupture, the surgical approach is still accepted as the standard for managing hydatid disease. The standard treatment is splenectomy (Hoffman, 1957) as Complete resection removes all parasitic and pericystic tissues.<sup>5</sup> During surgical treatment extreme caution must be taken to avoid rupture of the cyst.

Total splenectomy, partial splenectomy, cyst enucleation and unroofing with omentoplasty are the various preferred surgical techniques to treat splenic hydatid disease. The splenic cyst with adhesions or infiltrations to nearby organ should be treated by total splenectomy. However, if the splenic cyst is infected, have metastatic implantations to adjacent organs or the locations and size do not allow safe resection, conservative surgical techniques such as partial splenectomy, cyst enucleation, deroofting of the cyst with omentoplasty or external drainage may be used.<sup>6</sup> Many trials are usually made for conserving the spleen, so as to avoid overwhelming post splenectomy sepsis (OPSI).<sup>7</sup> Partial splenectomy carries a risk of poor vascular control when incising the splenic tissue while unroofing the cyst wall leaving behind a residual cavity carries the risk of postoperative infection. For the above reasons and the possibility of multiple splenic cysts, total splenectomy should be the method of choice, especially in the presence of a communication between the spleen and nearby organs, such as the stomach, colon and diaphragm. Laparoscopic approach has also been advocated for uncomplicated hydatid cyst of the spleen. Chemotherapy and newer methods, such as puncture, aspiration, injection, and re-aspiration (PAIR) technique using hypertonic saline or 0.5% silver nitrate solutions before opening the cavities tends to kill the daughter cysts. Medical treatment is the mainstay of treatment in the postoperative follow-up period. Anthelmintic drug therapy using Benzimidazole chemotherapy drugs with Albendazole 10–15 mg/kg/day for one month or Mebendazole 40–50 mg/kg/day for 3–6 months, in addition to Praziquantel 40 mg/kg/wk for 2 weeks pre and postoperative to reduce the chance of anaphylactic shock and decrease the tension in the cyst wall are used. We performed total splenectomy. After the splenectomy, the patient was given prophylactic vaccination against Streptococcus pneumoniae, Haemophilus influenza type b and Neisseria meningitidis, and was started on a six month course of prophylactic penicillin. No post-splenectomy infection was encountered. According to our knowledge, Primary Giant solitary hydatid cyst of spleen of size 250 × 200 mm containing fluid around 3.9 litres, the one of the first case to be reported case in the literature

Percutaneous drainage of uncomplicated hepatic hydatid cysts can be performed safely and results in the disappearance of the cyst. The efficacy of percutaneous drainage is similar to that of standard treatment with cystectomy, in terms of reducing the size of the

cyst and causing its disappearance over a period of up to two years. The advantages of percutaneous drainage include a shorter hospital stay and a lower complication rate.

Albendazole is an effective adjuvant therapy in the treatment of hydatid cyst. There are less chances of recurrence in patient who received albendazole therapy.

Gil-Grande et al. reported that albendazole sterilizes up to 72.3% of cysts by the end of the first month and 94% at the end of 3 months of treatment.

### 3. Conclusion

In conclusion, The infrequency with which it is encountered makes splenic hydatid disease a formidable early diagnostic challenge especially in nonendemic areas. Hydatid disease should be considered in the differential diagnosis of all cystic masses in the spleen/(**abdomen**), especially in the geographical regions where the disease is endemic. The splenic hydatid cyst may become a challenging surgical problem. Preoperative evaluation should be carried out carefully. Computerised tomography scan is the most sensitive investigation for diagnosis. The anatomical relations of splenic hydatid cyst should be demonstrated before surgery on account of varied presentations. Although the management must be individualized for each patient, a surgical resection is the best curative procedure. Postsurgical pharmacological treatment is necessary to ensure complete healing.

### Conflict of interest

None.

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None.

### Ethical approval

Yes we had taken written consent regarding publication.

### Author contributions

Dr. Mahesh M. Pukar – corresponding author who diagnosed, operated the patient and prepared manuscript of case report

Dr. Shabari M. Pukar – assisted for investigation, surgery and searching references.

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